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A school-based screening program for fetal alcohol syndrome ☆

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Abstract

Fetal alcohol syndrome (FAS) is a common cause of birth defects and neuropsychiatric impairment. Identification of affected people is crucial for early entry into intervention programs and for the development of prevalence estimates. The objective of this project was to determine if screening for FAS in a community elementary school-based setting was feasible, to estimate prevalence in the screened population, and to determine if a screening program for FAS can be implemented using available personnel from the community.

The FAS Screen was used to screen kindergarten students enrolled in a school system. Students with scores on the FAS Screen above the cutoff for a positive screen (20) were referred to one of several diagnostic clinics for evaluation.

Over a 9-year period, 1384 students were screened and 69 (5%) had a positive screen (20 or above). These 69 children were then seen in a genetics/dysmorphology diagnostic clinic and 7 (10%) were found to have FAS ($n=6$) or partial FAS ($n=1$). The prevalence of affected children (FAS and partial FAS) was 1 per 198 students or 4.3 per 1000.

The FAS Screen was completed annually by school staff, teachers, social workers, and psychologists. The test has acceptable epidemiologic performance characteristics in a community setting. The screening takes about 8–10 min. The procedure was well accepted in the community. This screening strategy was inexpensive to implement (less than US$8.00 per student), and can be easily included with the other screens completed at kindergarten entry.

Keywords: Fetal alcohol syndrome; Screening; Diagnosis; Schools; Cost; Prevalence

1. Introduction

Prenatal exposure to ethanol is the causal factor in fetal alcohol syndrome (FAS). FAS is a highly variable syndrome often presenting with variable neurological deficits or mental retardation [1,2]. Prevalence estimates for FAS range by nearly 100-fold [2,7,9]. Prevalence rates vary from 0.33 cases per 1000 live births to 19 per 100 [1,2].

Much of the variation appears to result from ascertainment strategies and active ascertainment nearly always produces higher rates than passive methods. Critical reviews of prevalence studies have reported that prevalence estimates developed with screening to identify at-risk populations nearly always produce higher prevalence estimates than those that do not have a screening step [4]. Screening also increases the efficient use of diagnostic resources and can be a low-cost strategy to identify children at low risk and at high risk [4,5]. Several screening strategies have been suggested (see Ref. [4] for a review).

Multiple screening gates were available to the researchers (pediatric clinics, birth defects clinics, developmental disorders clinics, Women Infant Child programs, Headstart, special education, school-based programs, women's alcohol treatment programs, criminal justice programs, juvenile justice programs, foster care or adoption programs). However, based on community input it was decided to use school-based settings for the screening. The strategy of developing the screening program in partnership with the community was an important concept that increased program ownership and we believe increases the potential for successful programs to be maintained.

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The purpose of this screening project was to examine the feasibility of screening for FAS in community settings. This project was felt to be a useful opportunity to accomplish three goals: (1) to examine the development of a gate to identify children who have development or behavioral disorders from prenatal alcohol exposure; (2) to estimate prevalence rates of FAS and to capture some cases of partial FAS; and (3) to examine the epidemiologic performance characteristics of the FAS Screen when applied by community personal in a community setting.

In this study, we utilized the FAS Screen, a rapid screening tool for community-based screening of FAS [5]. The tool was normed on children from North Dakota, Minnesota, and South Dakota.

2. Method

2.1. Screening tool development

The FAS Screen is a 32-item screening test (Appendix A). Development of the tool has included several revisions over the past 10 years to improve the weighing score for each of the items and to improve the performance of the screening tool. The 32 items used in the tool were developed from a item pool of over 60 items in the various revisions of the tool. Data on the epidemiologic performance characteristics of the FAS Screen have been presented [5] The sensitivity in the norming sample was 100%, the specificity was 94%, the positive predictive value was 92%, and the accuracy was 94%. Sensitivity (positive screen in subjects with disease) estimates how well the screening test identifies people who have the condition. Specificity (negative screen in subjects without the disease) estimates how the test correctly excludes people without the condition. Positive predictive value is the postscreening test probability that a person with a positive screen has the condition. Accuracy estimates what proportion of the screening results are correct.

Screening is not a brief diagnostic tool. As a result, some items important for diagnosis may not be included in a screening tool and some items rarely present in the diagnosis of individuals may be useful in screening. The development of efficient screening nearly always represents a choice between application of most of the diagnostic signs of a disorder which increases the technical difficulty of screening for a disorder like FAS and a choice to include a few broadly descriptive diagnostic signs which will increase the ease of utilization but decrease the sensitivity and specificity of the screening tool. Two useful examples are the inclusion of growth impairment and exclusion of short palpebral fissures in the screen. The FAS Screen captures all children with growth impairment not just those with growth impairment due to FAS. Short palpebral fissures, a sign of considerable interest to diagnosticians, is highly complex to measure. This is beyond the capabilities of nearly all community personnel. However, the measurement of growth is easily taught and can be performed by a large number of personnel in community settings. As a result, diagnosis signs like palpebral fissure length were not included in the FAS Screen. The screening tool is demonstrated in Appendix A.

2.2. Staff training to administer the screening

A 4-h screening training was completed in the community. The conceptual basis for screening was discussed. Each item of the screening tool was reviewed and demonstrated. The goal is to screen out low-risk children and identify a high-risk population. The FAS Screen in a community setting typically screens out as low risk about 94–96% of children [5].

2.3. Screening implementation

The screening project is supported by the school. The cost of diagnosis is billed to insurance or medical assistance. Some children are charged on a sliding fee scale. No child is refused due to inability to pay. The diagnostic clinics are held one to two times per year. Children who had scores above the cutoff or during the screening and miss the clinic appointment are then seen at one of two regional referral centers either 50 or 200 miles away in other identical genetic dysmorphology clinics.

Consent to participate was addressed and the school decided that a passive consent process was appropriate. A separate consent form for FAS screening was utilized and the parents of children enrolled were sent a form indicating that if they did not want their child screened they should return the form to the school indicating their desire not to have their child screened. The children who were not able to be screened may represent a high-risk group; however, the prevalence rate found in this study does suggest that many at-risk children were screened. We do not have data on those children who were not screened. Students are screened in the fall at the start of kindergarten. Screening takes about 8–11 min per child. Every child is screened even if they have a past diagnosis of FAS. This is important in a screening clinic where the epidemiologic performance characteristics of the tool (sensitivity, specificity, and accuracy) are of interest. If children with previously diagnosed FAS are not detected the tool

Table 1
Prevalence for screening population by year, number of positive screens (score of 20 or above), and diagnosed cases after evaluation by a medical geneticists with experience with FAS

Year	Number of children screened	Positive screen		FAS		Partial FAS	
		n	%	*n*	%	*n*	%
1992	132	11	8.3	2	1.5	–	
1993	126	7	5.6	–		–	
1994	120	6	5.0	–		–	
1995	144	9	6.2	–		–	
1996	197	8	4.1	2	1.0	–	
1997	195	7	3.6	1	0.51	1	0.51
1998	170	9	5.3	1	0.59	–	
1999	155	5	3.2	–		–	
2000	145	7	4.8	–		–	
Total	1384	69	5.0	6	0.43	1	0.072

would not be satisfactory as a screening tool for unidentified cases. A positive screening was defined as having a score of 20 or higher.

Children with a positive screen were referred to a local genetics dysmorphology clinic for evaluation. The school records, past medical records, and a maternal interview when the mother is available are also completed. Each child is seen for an individualized evaluation. The FASDC was used by the medical geneticist to record the signs of FAS or other genetic or dysmorphic syndromes [6]. The diagnostic criteria utilized were from Sokol and Clarren [8]. We also retrospectively applied the criteria from the Institute of Medicine Report on Fetal Alcohol Syndrome [8]. The final assessment report was sent to the child's physician and shared with the school to facilitate educational planning.

The community-specific prevalence of children who screened positive for FAS and those diagnosed with FAS or partial FAS was calculated.

3. Results

3.1. Prevalence

Data from nine consecutive years of screening were available and included in the study During the 9-year period, over 98% of the children enrolled in the school were screened. Of the 1384 children who were screened, 69 (5%) had a positive screen (score of 20 or above). The annual prevalence of a positive screen in those children who were screened ranged from 3.2% to 8.3% over the 9 years.

These children were then sent to a genetics/dysmorphology clinic for evaluation. Of this group, 7 of 69 (11%) were diagnosed with FAS (6 of 69; 8.6%) or partial FAS (1 of 69; 1.4%) as shown in Table 1. Each child diagnosed with FAS also met the criteria from the Institute of Medicine criteria when applied retrospectively by chart review (six FAS with confirmed maternal exposure and one partial FAS with confirmed maternal exposure) [8]. The prevalence of FAS was 1 per 230 kindergarten students or 4.4 per 1000.

3.2. Reliability

Using this data, community-specific estimates for sensitivity and specificity values for the screening tool were calculated (Table 2). The sensitivity of this test was 100% (no children in the sample who screened negative had a previous diagnosis of FAS or partial FAS). The specificity was 95.43% (4.5% of the children had a positive screen but did not have FAS). The accuracy rate of the screening tool was 95% (95% of the children were accurately categorized with the screening tool).

Table 2
Epidemiologic performance characteristics of the FAS Screen for 1384 children in the community

	FAS		Not FAS		Sensitivity	Specificity	Accuracy
	n	%	*n*	%			
Screened Positive	6	8.7	63	91.3	100	95.43	95.44
Screened Negative	0	*	1315	100			

* Based on a separate evaluation of the sample by a different program.

4. Discussion

This project demonstrates efficiency effectiveness and efficiency of a community-based screening strategy for FAS. The community-based utilization of the FAS Screen is time efficient (takes less than 15 min per child) and produces a small population for referral (5%). Of the population with a positive screen, 8.7% had a diagnosis of FAS. The tool has acceptable epidemiologic performance characteristics in this community setting (Table 2). These values are very similar to the performance parameters established during the normative process [5]. The costs were are quite similar to the estimates published previously [4,5].

The screening of early school populations facilitates early identification. Early identification may enhance prevention of secondary disabilities in the affected child.

In Phase 2 of this project, a supplemented screen for a more common outcome from prenatal alcohol exposure is being considered for inclusion to the screening protocol This tool will screen for alcohol-related neurodevelopmental disorders (ARND) [9]. The ARND screen has been developed for community-based screening for ARND [3,4]. The population prevalence of ARND and alcohol-related birth defects (ARBD) are estimated to be four to five times more frequent than FAS [5,7]. If these estimates are supported by future research the prevalence of FAS, ARND, or ARBD could range from 1 in 46 to 1 in 56 children in this community [5]. The total prevalence of FAS and related disorders could be anticipated to range from 17.6 per 1000 to 22 per 1000 students. Since ARND may be much more prevalent than FAS the development of an acceptable screening tool for ARND has the potential to be very useful.

The school completes the FAS screening without additional financial, logistical, or technical support. The model developed in this community may be useful in other community settings. The ease of implementation combined with the low cost of the screening program offers schools or other community organizations an opportunity to identify children with FAS. Additional community-based projects using the FAS Screen are underway in Headstart programs, Early Periodic Screening Diagnostic Testing, and in a county social services program. The results reported here are similar to a previous 1-year screening program in the Early Periodic Screening Diagnostic Testing program in North Dakota, where 2800 children were screened and 28 cases of FAS-ARND that were previously undiagnosed were identified.

Further research using a variety of screening strategies in a variety of settings are required. Screening may be a useful strategy for communities to utilize in developing an FAS intervention program.

Appendix A

Larry Burd, Ph.D.

FAS SCREEN FORM

Name _____ DOB___/___/___ AGE _____ SEX (circle one) F M DATE OF EXAM __/__/__

CHILD S RACE (circle one)	HEIGHT_____ INCHES	<5%	Y____ N____	10
1) white	WEIGHT_____POUNDS	<5%	Y____ N____	10
2) NA	HEAD CIR._____CM	<5%	Y____ N____	10
3) other				

HEAD AND FACE	EARS STICK OUT (Protruding Auricles)	Y____ N____	4
	SKIN FOLDS NEAR INNER EYE (Epicanthal Folds)	Y____ N____	5
	DROOPING OF EYELIDS (Ptosis)	Y____ N____	4
	CROSS-EYES, ONE OR BOTH EYES (Strabismus)	Y____ N____	3
	FLAT MIDFACE/CHEEKS (Hypoplastic Maxilla)	Y____ N____	7
	FLAT/LOW NOSE BETWEEN EYES (Low Nasal Bridge)	Y____ N____	2
	UPTURNED NOSE	Y____ N____	5
	GROOVE BETWEEN LIP AND NOSE ABSENT OR SHALLOW	Y____ N____	5
	(Flat Philtrum)	Y____ N____	4
	THIN UPPER LIP	Y____ N____	4
	CLEFT LIP OR CLEFT OF ROOF OF MOUTH (present or repaired)		
NECK AND BACK	SHORT, BROAD NECK	Y____ N____	4
	CURVATURE OF THE SPINE (Scoliosis)	Y____ N____	1
	SPINA BIFIDA (History of Neural Tube Defect)	Y____ N____	4
ARMS AND HANDS	FINGERS, ELBOWS (Limited Joint Mobility)	Y____ N____	4
	PERMANENTLY CURVED, SMALL FINGERS, ESPECIALLY		
	PINKIES (Clinomicrodactyly)	Y____ N____	1
	DEEP OR ACCENTUATED PALMAR CREASES	Y____ N____	4
	SMALL NAILS/NAIL BEDS (Hypoplastic Nails)	Y____ N____	1
	TREMULOUS, POOR FINGER AGILITY (Fine Motor Dysfunction)	Y____ N____	1
CHEST	SUNKEN CHEST (Pectus Excavatum) OPTIONAL	Y____ N____	3
	CHEST STICKS OUT (Pectus Carinatum)	Y____ N____	1
	HISTORY OF HEART MURMUR OR ANY HEART DEFECT	Y____ N____	4
SKIN	RAISED RED BIRTHMARKS (Capillary Hemangiomas)	Y____ N____	4
	GREATER THAN NORMAL BODY HAIR,HAIR ALSO ON		
	FOREHEAD AND BACK (Hirsutism)	Y____ N____	1
DEVELOP MENT	MILD TO MODERATE MENTAL RETARDATION (IQ<70)	Y____ N____	10
	SPEECH AND LANGUAGE DELAYS	Y____ N____	2
	HEARING PROBLEMS	Y____ N____	1
	VISION PROBLEMS	Y____ N____	1
	ATTENTION CONCENTRATION PROBLEMS	Y____ N____	2
	HYPERACTIVITY	Y____ N____	5

COMMENTS:

SCORE TOTAL_____
Refer if 20 or above

For additional forms or information on FAS, FAE or ARNDD contact:

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